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COWPER'S DUCT CYST - AN UNUSUAL PRESENTATION OF LOWER URINARY TRACT OBSTRUCTION IN PAEDIATRIC AGE GROUP.

Paediatric Surgery

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KEYWORDS

INTRODUCTION

Bulbourethral or Cowper's gland is a paired pea sized gland derived from the pericloacal mesoderm under the influence of androgen. They are located in wide base of external urethral sphincter muscle in the deep perineal pouch, posterolateral to membranous urethra and superior to the perineal membrane. The ducts open into the bulbous urethra The glands secrete an alkaline mucinous secretion containing glycoprotein which lubricates the urethral passage before sperm ejaculation. [1] Cystic dilatation of gland or syringocele is secondary to duct ectasia or distal duct obstruction. It is commonly seen in the paediatric age group. It may be asymptomatic or present with urinary complaints. Diagnosis is based on imaging and endoscopy findings. Management is predominantly by endoscopic intervention.

Case Report

A 12-year boy presented with intermittent, dull aching pain in left upper abdomen since 2 years. He was under treatment for constipation. There was no history of fever, abdominal distension, vomiting, dysuria, hematuria or pyuria. Abdomen and genitalia were normal. There was fecal loading on per rectal examination.

X- Ray showed fecal loading in distal colon. Ultrasonography revealed left renal AP diameter of 1.2cm without hydroureter with normal contralateral kidney and ureter.

He was prescribed stool softener along with dietary advice. At 6 weeks follow up, stool pattern was normal. However, dull aching abdominal pain was persistent. There was also persistent hydronephrosis on ultrasonography. Voiding cystourethrogram showed evidence of Grade III bilateral vesicoureteric reflux (VUR). A small radiolucent structure was seen in the region of the proximal penile urethra(Fig.1).

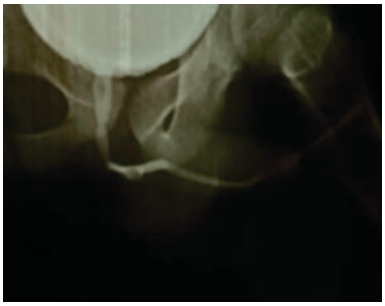

Fig.1

EC scan and DMSA scan were normal. On cystoscopy, a cystic, mucosa lined, approx. 2cm*0.5cm structure partially occluding the lumen was visualized in the proximal anterior urethra (Fig. 2).

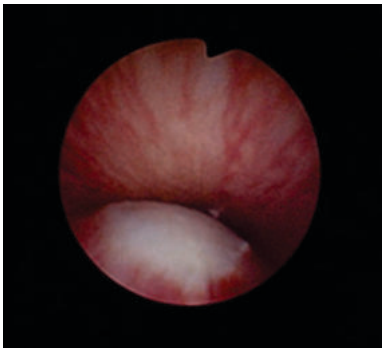

Fig. 2

The posterior urethra, verumontanum and bladder neck were normal. Bladder was mildly trabeculae with normally placed bilateral ureteric orifices. The cystic structure was incised. DE roofing of cyst was done resulting in improved caliber of urethral passage. Patient was catheterized and kept on antibiotic coverage.MCU done at 6 weeks showed no urethral lesion. VUR was persistent. Check cystoscopy showed resolution of Cowper's duct cyst without any significant scarring. (Fig.3)

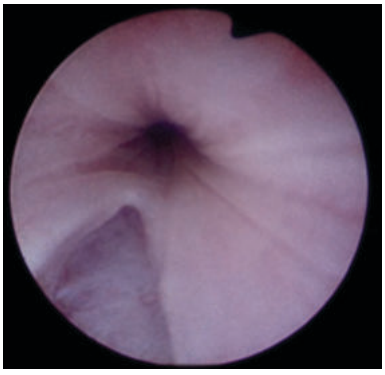

Fig. 3

DISCUSSION

Cowper's duct cyst i.e. syringocele was first described in 1881 and literature with classification was given by Maizel et al in 1983[2]. It can be congenital or acquired. It is commonly seen in paediatric population than in adults. In acquired cases, it is usually secondary to trauma, prolonged catheterization or infection. Stasis and pressure changes leads to obstruction of the duct orifice leading to accumulation of mucus and cystic dilatation.

Maizel et al classified syringocele based on clinical symptoms and radiological findings. [2]

1) Simple syringocele- minimally dilated duct
2) Perforated syringocele- diverticulum like connection with urethra
3) Imperforate syringocele – submucosal cyst
4) Ruptured syringocele – thin membrane following ruptured cyst

Campobasso et al in 1996 gave classification as follow [3] –
1) Obstructing type 2) Non obstructing with hematuria, incontinence, fever, UTI

Beaver et al recommended a classification based on duct opening in urethra; open variety has a continuous lumen between the urethra and duct presenting as urinary incontinence while closed one presents as obstructive urinary symptoms [4].

Severe cases in children can lead to bilateral hydroureteronephrosis, vesicoureteric reflux or scrotal abscess while in adults they may present as discharging perineal sinuses or fistulas. [5-7] Imaging modalities play major role in diagnosis of syringocele.

Transrectal and transperineal ultrasonography can detect Cowper's gland cystic lesion, the drawback being insufficient tissue penetration in penile bulb.

Micturating cystourethrography is gold standard in diagnosing syringocele communicating with urethra. Cystourethroscopy shows an imperforate syringocele as an extrinsic mass arising from the bulbar urethral floor, while a perforated syringocele would manifest as a pouch connecting to the bulbous urethra. MRI being radiation free and noninvasive imaging with broader field of vision can be used to diagnose syringocele by exhibiting homogeneous cystic lesion. [8]

Based on history, clinical examination and imaging modalities, syringocele can be differentiated from urethral duplication, anterior urethral valve, anterior urethral diverticulum, congenital narrowing of bulbar urethra, urethral stricture, megalourethra, periurethral abscess, perianal abscess, congenital urethral folds, prolapsed posterior urethral valve and diverticulum [9].

Asymptomatic syringocele can be managed conservatively but symptomatic one needs intervention as spontaneous resolution is not possible. Cystoscopic intervention by deroofing or marsupialization by using hook diathermy / electrocautery/ scissor is the preferred treatment for symptomatic syringocele. A typical, efficient method of marsupialization for open and closed syrinx is to unroof the cyst by disconnecting it from the urethra.

Bevers et al have proven the effectiveness of endoscopic deroofing. The successful implication of the Holmium: Yttrium Aluminum Garnet (YAG) laser for syringocele intervention which Piedrahita and Palmer initially proposed, is now more widely practiced, and recent case reports depicted that there is no need for urethral catheterization after this treatment [10].

Failed endoscopic approach can be managed with transperineal open approach by ligating Cowper's duct [6]. Syringocele with big diverticulum can be treated with diverticulectomy.

CONCLUSION

Although Cowper's duct cyst is a rare entity, it should be considered as one of the differentials for symptoms of obstructing uropathy. Radiological and endoscopic evaluation plays a major role in diagnosis and identifying the severity of its presentation. Endoscopic intervention is gold standard treatment modality with good outcome.

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